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Case Report

Urinary Bladder Xanthoma: A New Case Report in Association with Resected Urothelial Neoplasm

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Abstract

Urinary Bladder Xanthoma (UBX) is an infrequent lesion that has been very rarely referred in medical literature. We described a case of UBX occurring subsequent to resection of a papillary urothelial neoplasm of low malignant potential. The patient is an 82-year-old man with arterial hypertonia and long-term hypercholesterolemia, presented for control bladder exam 2 years after transurethral bladder resection.

The histologic diagnosis was made by the presence in lamina propria of lipid-laden macrophages, CD68+ immunohistochemically. In vicinity of the UBX, in a histologic context of oedema and congestion, a dense cicatricial fibrosis was observed in the bladder wall.

A few case studies of post-local surgery cicatricial bladder-wall fibrosis in association with UBX have shown. The morpho- and pathogenesis of UBX are discussed.

Keywords: Urinary bladder; Xanthoma

Introduction

Urinary Bladder Xanthoma (UBX) is almost always an incidental finding and represent a characteristic yellow colour lesion on cystoscopy with pathognomonic features on histopathology [1]. Microscopically, UBX is a localized non-neoplastic accumulation of lamina propria mucosae histiocytes containing lipids (foamy macrophages). The submucosal location of the UBX explain the presence of gross lesion during the cystoscopy [2].

UBX is rare with only 19 cases (including our case) previously reported worldwide [1,3,4]. It could be an isolated UBX but the lesion is frequently associated with urothelial neoplasm, often of low malignant potential [4].

Although already described by Scholl in 1945, the etiology of UBX still remains unclear.

Case Report

An 82-year-old man presented for control bladder exam 2 years after Transurethral Resection (TUR) of bladder papillary urothelial neoplasm of low malignant potential. His medical history included arterial hypertension (195/80 mmHg), gonarthrosis, chronic gastritis with intestinal metaplasia, *Helicobacter pylori* +. There were no significant lower urinary tract symptoms. Physical examination was unremarkable with no lymphadenopathy. Urinary cytology showed no malignant cells and culture failed to grow any organisms. Ultrasound scan of the kidney was normal. A flexible cystoscopy showed an abnormal yellow-white solitary plaque up to 11 mm in maximum diameter on the lateral left side of the bladder. Adjacent to the actual lesion on the posterolateral left side of the bladder, the cicatrix of the precedent bladder TUR measured 7mm, was observed.

Blood counts and other haematological investigations were, otherwise, within limits. His lipid profile showed total cholesterol 270 mg/dL, tryglycerides 280 mg/dL, high-density lipoprotein 35 mg/dL and low- density lipoprotein 180 mg/dL. The resected material included 4 tissue fragments measured 3 - 6 mm.

Histological examination showed aggregates of foamy histiocytes with abundant vacuolised cytoplasm and dense round

nuclei in lamina propria (Figure 1a). These cells showed strong immunoreactivity with CD68 (Figure 1b) and were negative for cytokeratin AE1/AE3, CD1a, PS100, periodic acid -Schiff reaction, von Kossa and Perls, and all special histochemical stains excluded infections (data not shown). Neither giant multinuclear cells nor cytoplasmic inclusions were observed which excluded both major entities in differential diagnosis: xanthogranulomatous cystitis and malakoplakia [2]. In one of the fragments directly beneath lamina propria containing foamy macrophages, is seen dissociation of superficial bladder musculature (muscularis mucosae and the outer part of detrusor muscle) by dense cicatricial fibrosis, well distinguishable and stained in yellow with hematoxylin-eosin-saffron staining (Figure 2a). Adjacent tissue is highly swollen and congestive with dilated capillaries (Figures1a and 2b). There was neither any significant inflammation nor evidence of malignancy. Treatment consisted of local excision. The follow-up cystoscopy at 6-month interval was normal.

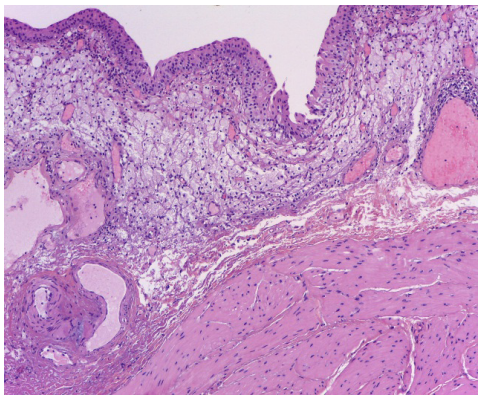

Figure 1a: Photomicrograph shows normal urothelium and lamina propria that contains clusters of foamy cells. Note: Dilated blood capillaries and venules directly beneath the lesion (hematoxylin-phloxine-saffron; x 100).

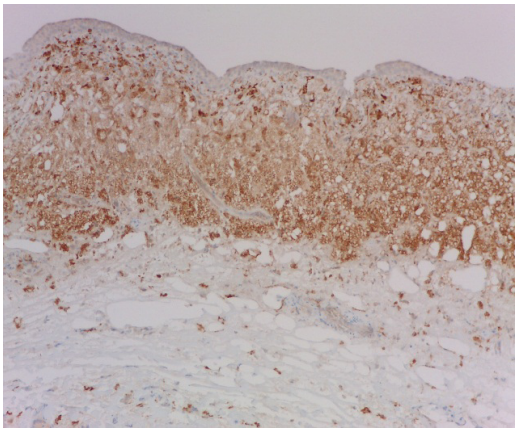

Figure 1b: Immunohistochemical staining of foamy cells with CD68, a macrophage marker (CD68 immunostain; x 100).

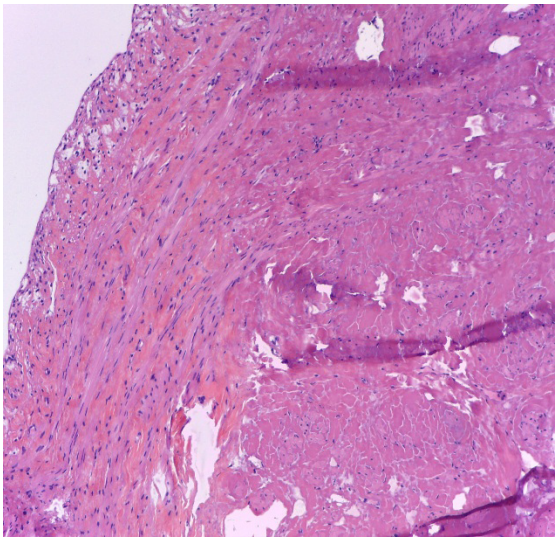

Figure 2a: Photomicrograph shows clusters of foamy cells in the lamina propria and cicatricial fibrosis between smooth muscle fibres (hematoxylin-phloxine-saffron; x 100).

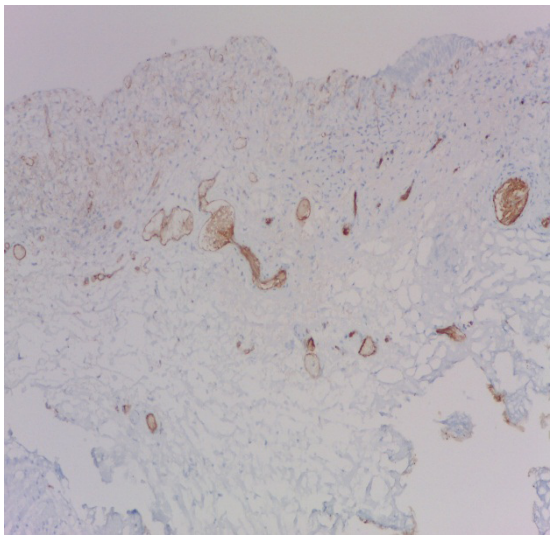

Figure 2b: The surrounding tissue is highly swollen and congestive, with dilated capillaries, whose wall is underlined with CD31, an endothelial marker (CD31 immunostain; x 100).

Discussion

UBX is a distinctive, very rare clinical and histopathological entity that should be considered. UBX is a non-neoplastic, reactive tumour-like process representing an incidental finding during a procedure or the course of investigation on the lower urinary tract. Although his clinical significance is unclear, they are important lesions because they may be confused with malignant proliferations.

Only 19 cases (including our case) were reported in the medical literature [1-4]. UBX are reported in older patients who present with nonspecific symptoms or hematuria and often have associated lipid anomalies [1-6]. When in association with urothelial neoplasm, the mean age of patients with UBX was 62.5 years (range: 51-69 years) and all were males [4]. Our patient is 82 years old male.

UBX may occur in either primary or secondary hyperlipidaemia or/and hyperholesterolaemia and these are more common cases of isolated xanthoma [2,4]. 5/6 patients with metabolic abnormalities had hyperholesterolaemia [4], as in the case presented by us.

The diagnosis is histological, based on non-neoplastic accumulation of lamina propria mucosae histiocytes containing lipids (foamy macrophages). The etiology, morpho-and pathogenesis of UBX still remain unclear. They are likely to be a result of an exaggerated accumulation of lipid-laden macrophages in response to global or/and local metabolic disturbances in ageing bladder mucosa.

The coexistance of UBX and bladder carcinoma has been documented in the literature [3,4]. In the largest published series of cases - 17 UBX, 11 (65%) originated within or adjacent to urothelial neoplasm [4]. When in association with urotelial neoplasm, UBX occur often in normolipidaemic states [4].

Some authors pay attention to the fact that UBX develop adjacent to urothelial carcinoma [7], but the pathogenesis of peri-tumoral xanthoma has not yet been established. Like gastric xanthoma [8], it may be speculated that increased release of oxygen free radicals in peri-tumoral area, may be involved in the formation of UBX.

There are single observations presented an UBX occuring subsequently to several local resections of urothelial neoplasm, trauma and inflammation [9,10]. To the best of our knowledge, we report the first histological observation of UBX in association with post-TUR mural bladder cicatrix. In our case we present a combination of metabolic disorders and resected urothelial neoplasm in old patient.

Similar observations are described in stomach xantomas, associated with gastrointestinal anastomosis: the lesion was near to the anastomosis and it is probably due to local disturbances in mucosal lipid metabolism [11]. It can be speculated that UBX may reflect the long duration and the importance of tissue volume defect, subsequently to previous local bladder-wall surgery.

Obviously when patients had lipid metabolic background disorders the process would be more easily achieved. Our observation is similar. We established a combination between general background (lipid profile metabolic disturbances) and local (mechanical disturbances in blood and lymphatic drainage) factors.

In conclusion, UBX is a benign condition with no premalignant potential. UBX is a rare, non-neoplastic, reactive tumour-like process, able to develop in old patients with impaired lipid profile nearby mural cicatrix from a previous tumour surgery. The addition of our case to the literature offers new morphogenetic data and reasoning useful for better defining the pathogenesis of this lesion.

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